

Pediatrics

Treatment of a high output nephrocutaneous urine leak following treatment of a giant calyceal diverticulum in a child

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ABSTRACT

Calyceal diverticula are non-secretory transitional-epithelium lined cavities that are connected to the collecting system through a small ostium. They are rarely seen in young children. Although most remain asymptomatic, the presence of stagnant urine can result in infection, stone formation and pain. Treatment may consist of percutaneous ablation or open surgical excision. Failure of the ostium to completely seal can result in a persistent leak. We present a case of a massive calyceal diverticulum in a child who developed a postsurgical nephrocutaneous fistula recalcitrant to conservative management that was successfully managed with injection of fibrin glue.

Introduction

Calyceal diverticula are congenital transitional-epithelium lined outpouchings that emanate from the renal calyx or pelvis and project in to the renal cortex. Although rarely encountered in children, when they do occur they can result in symptoms of flank pain, urinary tract infection, calculus formation and/or hematuria.^{1,2} Treatment of symptomatic lesions mirrors that seen in adults with percutaneous approaches being utilized for smaller diverticula and open surgery/robotic surgery reserved for the very large. Although attempts are made to completely close the ostium, leaks do occasionally occur in the post-operative setting. We report on a minimally invasive approach for managing a persistent leak following initial treatment of a 22 cm calyceal diverticulum.

Case report

A 10-year-old boy presented with flank pain and gross hematuria after sustaining a flank injury while playing American style football. A CT demonstrated a giant mass measuring 22 cm in diameter involving the left kidney (Fig. 1a and b). The differential diagnosis consisted of a lower pole UPJ obstruction, a large simple cyst, and a calyceal diverticulum. Due to its large size, a decision was made to first decompress with a percutaneous drain. Contrast injection of the cyst demonstrated findings consistent with a calyceal diverticulum and an ostium that extended to the renal collecting system. At the time of open retroperitoneal resection a 3mm diameter ostium was identified. Management

consisted of fulgurating and over sewing the ostium after which neighboring parenchyma and renal capsule were used to cover the site. A percutaneous Jackson Pratt drain and ureteral stent were left in place following the procedure. High output drainage (>250 ml/day) was noted through the JP drain. The creatinine level of the fluid was consistent with urine. The child was sent home on postoperative day 10. High output persisted for four more weeks. A retrograde pyelogram readily demonstrated the site of extravasation (Fig. 2). Subsequent management consisted of up-sizing the stent from 5 to 8 French and, in attempt to minimize retrograde passage of urine from the bladder, a suprapubic cystostomy drain was later placed. These maneuvers proved fruitless in resolving the leak. After 6 additional weeks of unsuccessful management, resolution of the high output leak was realized by removing the Jackson Pratt drain and injecting Tisseel® fibrin sealant (Baxter International, Deerfield, Illinois, USA) through the mature fistula tract. Drainage immediately resolved and the ureteral stent was subsequently removed. Follow-up studies have demonstrated no further evidence of leak or cyst. The child has been free of infection and has normal imaging 5 years following the procedure (Fig. 3).

Discussion

Calyceal diverticula are cavities within the renal parenchyma that are typically found in the polar regions of the kidney. They are believed to form due either to persistence of late ureteral bud divisions that fail to regress or to rupture of a simple renal cyst in to the collecting system. They are non-secretory and most remain asymptomatic. However, due

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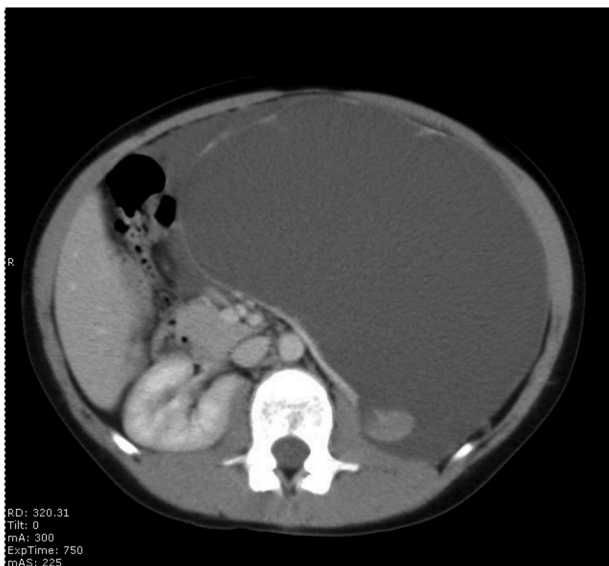


Fig. 1a. Axial CT image demonstrating giant left sided calyceal diverticulum.



Fig. 1b. Coronal CT image demonstrating giant left sided calyceal diverticulum.

to their non-secretory nature, urine can remain stagnant and result in infection, stone formation and pain. Symptomatic calyceal diverticula are rarely seen in children. When they do come to attention surgical options range from percutaneous ablation of the structure to open and robotic surgical excision. We favor percutaneous ablation/coagulation of the inner aspect of the diverticulum and have noted a high success rate with this technique. However, we felt that in the present patient the



Fig. 2. Retrograde pyelogram demonstrating high volume leak from site of resected calyceal diverticulum.



Fig. 3. Renal Ultrasound of affected kidney 5 years following surgical removal of giant calyceal diverticulum.

enormous size of the diverticulum made successful percutaneous ablation of the inner lining improbable. We therefore elected to perform open excision through a retroperitoneal approach.

Persistent leaks from the kidney to the skin can occur following partial nephrectomy.³ Nephrocutaneous fistulas have also been noted after penetrating trauma, as a result of xanthogranulomatous pyelonephritis, tuberculosis infection and obstructing stones. Most postsurgical fistulas resolve with conservative management of observation, ureteral stenting and/or bladder drainage. In our patient these measures did not prove successful in resolving the problem.

Fibrin glue is a topical biological adhesive that imitates the final stages of coagulation. It consists of a solution of concentrated human fibrinogen, which is activated by the addition of bovine thrombin and calcium chloride. Since these sealants first entered the US market in

1998, numerous uses have been identified in nearly every organ system. Fibrin glue has been used extensively in management of renal trauma and to control bleeding and prevent leakage following partial nephrectomy. Treatment of urinary fistulas has been treated in a number of adult conditions.⁴ Fibrin glue has also been proposed as a means of sealing a percutaneous tract and thereby performing “tubeless” percutaneous nephrostolithomy.⁵

Our pediatric patient underwent open surgical excision of the giant diverticulum. Despite careful excision, direct closure of the calyceal ostium and multiple layer coverage with neighboring parenchyma and renal capsule, a leak occurred that was resistant to conservative measures. The simple instillation of fibrin glue at the time of drain removal provided a quick and durable resolution of this complication. Although we enthusiastically endorse this strategy for resolving a persistent nephrocuteaneous leak, care should be taken when instilling the fibrin

sealant so as not to inject directly in to the renal collecting system since this could result in obstruction of the collecting system.

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